Factors associated with Pain in Adolescents with bilateral Cerebral Palsy.

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Abstract

Aim: Pain is increasingly recognised as an important symptom to be addressed as part of clinical care in people with cerebral palsy (CP). We explore factors associated with pain and pain severity in a cross-sectional study of a population cohort of young people (YP) with bilateral CP and compare parent/carer (PC) and YP self report.

Method: Of the 278 survivors aged 13-19 years from the South Thames region, SH&PE cohort, of 338 YP with bilateral CP, 212 PC and 153 YP completed questionnaires on the presence, severity, timing, site, associated factors, impact and treatment of pain.

Results: Seventy percent of PCs reported pain occurring in the last three months, 59% within the last week and 50% within the last day associated with severity of motor

impairment and ambulant/ non- ambulant and 'regularly experienced' by 56%. Of YP able to do so, 63% reported pain in the last three months, 50% the last week and 42% the last day, with 48% reporting regular pain. There was strong agreement between PC and YP reporting of pain and its severity over the last 3 months. Pain severity was associated with severity of motor impairment, health problems, particularly constipation, spasticity, greater equipment use and higher emotional score but not gender, intellectual disability, speech level or maternal education. Sites of pain were arm, leg, back, chest and hips, multiple in two-thirds. Pain in individuals with less severe motor impairment was associated with voluntary movement and with being moved in those with a severe motor impairment. Greater pain severity at all time periods had a negative impact on PC and YP measures of quality of life in both physical and psychological well-being.

Interpretation Increasing awareness of the co-morbidities in CP, particularly general health and constipation, may help more effectively treat and thereby reduce the high levels of pain experienced by young people with cerebral palsy

Word count Abstract 305

Word count 4034

What this study adds

- Regular moderate or severe pain is reported in YP with bilateral CP at all levels of GMFCS but more frequently in YP who are non-ambulant compared with ambulant.
- General ill health and particularly constipation is strongly associated with severity of pain after controlling for severity of CP.
- Pain occurs most often in ambulant YP during voluntary activity and in YP who are non-ambulant when being moved.
- There is strong agreement between PCs and YP about pain presence and severity.

Introduction

Cerebral palsy is usually thought of as a disorder of motor function but multiple co-morbidities impact negatively on other body functions (NICE 2017)[1]. Pain has been increasingly recognised as both common and negatively affecting quality of life and both participation and 'well-being' for children and young people [2-6]. Prevalence rates of pain in young people with CP vary in recent studies from 27% to 77%. Such differences depend on a number of factors[6-8]: the age range (pain increases with age[6] both in prevalence and intensity[5,8,]); gender (pain is reported more frequently in females[6], increasingly so with age);

the population (whether population based cohorts[3,4], a total population of CP [5,6], or clinical populations[8]) and whether non ambulant[5,] or non-verbal[8] ; the specified recall time (no time frame as in Westbon 37%[7] or a time frame of the last week and the last four weeks, 60% and 73% respectively[3,4]. Whether intensity of pain is reported is also relevant eg in 101 non-verbal children and adolescents with CP aged 2 to 20 years parent/carers (PCs) reported pain in 65% during the last 4 weeks; pain was intense in 17%, and daily pain occurred in 28%[8]. Pain experience may change over time as may intensity[8]. Those who at a younger age have no pain may develop pain and those with more severe initial pain and higher gross motor function have lower pain at follow-up[9]. Also relevant to prevalence is who is reporting the pain: self-report or physician observer, 54% versus 39% respectively[10]. Parent and self-report, where both can contribute, have been found to be moderately correlated in 13-17 year olds although parents tended to relatively overestimate their child's pain if self-reported pain was infrequent or mild and underestimate it if self-reported pain was frequent or severe[4].

Factors associated with pain are severity of CP, distribution of CP (4 limb CP having more severe pain) and motor type (dystonia) in one study[8] although not in Westbom et al[7]. Reports of pain are not associated with socioeconomic factors, including parental education in two large studies[3,4,5]. Musculo-skeletal causes are assumed to be most common[11,12,13] but constipation is noted [8, 10] . Sites of pain are most commonly hips, knees and feet[13]. Greater levels of pain have been positively associated with emotional difficulties in YP [4] who experience both increased pain intensity and pain anxiety[14]. Pain has been linked explicitly to therapy and clinical procedures[4] such as stretching procedures[15] (in the latter study parent/carers underreported pain compared with self and physio report).

Systematic enquiry about pain from PCs and YP is now part of good clinical care in CP. Ascertaining pain in individuals who can self-report is relatively straightforward. Ascertaining pain in the non-verbal population is more problematic as pain has to be inferred from behaviour and relies on parent/carer/observer report. High agreement was reported between physiotherapist and parents in non-verbal children[9]. Which pain questionnaire to use has varied across studies and a systematic review concluded that tools were often not validated in children with CP nor was there one tool to meet the needs of all children experiencing chronic pain [16].

In a recent commentary in Developmental Medicine and Child Neurology Ramstad[17] posed the question, "do we need another report on pain in cerebral palsy?" However clinically important questions remain. Pain is discussed in the literature as largely musculo-skeletal in origin. We predicted that other factors such as gastrointestinal dysmotility, especially constipation, were important and potentially amenable to treatment. Pain has also been

explicitly linked to therapeutic interventions which has important implications for all clinicians. We predicted that pain is linked to different times and activities in different groups and possibly to the use of orthoses and postural equipment. We wanted to assess whether PCs and YP were in agreement about presence, timing, site and severity of pain- a clinically important assumption for those who have limited communication. We were interested in what treatments individuals use to control pain as Westbom found that in clinical practice, although structured assessments had captured the presence of pain, very little intervention was recorded in the notes of the YP[7]. The implications of pharmacological treatment in Chronic Pain in Cerebral Palsy was also highlighted in the recent review on pathophysiology by Blackman et al[18]. To address these questions we report the presence, severity, timing, site, impact, associated features and self-management of pain in a cross-sectional study of a population-based cohort of YP with bilateral CP.

Methods

Population The cohort is derived from a total population of children with bilateral cerebral palsy (CP) born 1989 to 1992 (inclusive) to mothers resident at the time of birth in a geographically defined area of south-east England (205,958 live births) who participated in a study of hip development in cerebral palsy; the Study of Hips and Physical Experience (SH&PE) [19,20]. As the original study focused on the development of hips uninfluenced by typical walking experience, the upper age limit for acquisition of CP was 15 months and children who died before 12 months of age were excluded. The cohort was examined at 2, 5 years and 13-19 years. See flow chart. Figure 1. This paper provides a cross-sectional analysis of the teenage assessment.

Measures

Motor. Functional motor ability was recorded using the Gross Motor Function Classification System (GMFCS I-V) (Palisano[21]) and separately as ambulant/non-ambulant (ambulant meaning any mobility using the legs with or without a walker/rollator etc). The passive range of movement was assessed using a goniometer. A total deformity score for the spine and lower limbs was calculated (see appendix). Type of motor disorder was recorded as predominantly spastic, dystonic, dyskinetic or other including ataxia. Current use of postural therapeutic equipment was recorded: standing frame, night lying support, spinal brace, hip abduction brace (total body), AFO and knee gaiters (limb) or none.

Evaluation of Pain. Presence and severity of pain was recorded on a visual Likert scale scored from 0 (none) to 10 (severe) pain over the preceding day, week and last 3 months; whether pain was regularly experienced and of what severity (mild/mod/severe), where (head, chest, back, arms, hips, legs, feet & tummy or other), (see appendix), when (at rest, when moving, when moved, when eating, daytime, at night or all the time), and when it was

worst. YP (and PCs) were asked about what treatment, if any, they used for pain (any analgesics used for regular pain were totalled for analysis purposes) and frequency of use (never/rarely; sometimes/quite often or a lot/all the time).

Impact of Pain. The explicit impact of pain on daily life was reported by YP using the Brief Pain Inventory a 10 sub-section questionnaire of the impact of pain on recreational activity, school/college/work, mobility, self-care, sleep, mood and overall enjoyment, scored 0 (none) – 3 (a lot) with a total score of 0-30) [22].

Quality of life was assessed using Kidscreen, a PC and YP reported 52 item questionnaire covering 10 dimensions (1-5 scale) physical health, psychological well-being in mood and emotions, self-perception, autonomy, home life, money matters, friends, school & learning and bullying in the last week[23].

General Health in the past 5 years was recorded in face to face interview with PCs and scored as 2 (definite), 1 occasional), 0 (none) for each of chest problems, epilepsy, gastrointestinal problems (including constipation) and 'other' (a mix of problems with shunts, urinary tract infection and other conditions but excluding orthopaedic procedures) giving a possible score of 8; in sub-analysis constipation was separately analysed.

Neurodevelopment and Behaviour

PCs reported communication and learning impairment as none, mild/moderate, severe. Speech was assessed directly by a research physiotherapist using the Pennington scale[24] (a 5 item scale from easily intelligible to no intelligibility and highly correlated with parental report of communication[25]). Children were categorised as having no Intellectual Disability-ID (0), mild/mod ID (1) or severe/profound ID (2) on the basis of the British Picture Vocabulary Scale (BPVS) score and additional school information. PCs completed the Strengths and Difficulties Questionnaire (SDQ)[26].

Statistical analyses were carried out using Stata 14 [27]. PC report informed the main analysis for presence, severity and associated features of pain because some YP could not self-report. As main outcomes we analysed the number with reported pain and pain severity in three different time periods and the presence of regular pain of moderate and severe degree. Pain severity scores (from the 11-point Likert scales) for the last day, week and 3 months were treated as continuous variables and absence/presence of regular moderate-severe pain was treated as binary. Factors considered as potentially being associated with the main outcomes included: severity of motor impairment, as measured by GMFCS level (continuous variable) or an indicator for ambulatory/non-ambulatory

(categorical); degree of joint deformity, as measured by deformity score (continuous); motor disorder type (categorical); use of postural modifying equipment (categorical); general health index (continuous) and separately constipation (categorical). Associations between these factors and PC report of pain were first explored in bivariate analyses. SDQ total score, SDQ emotional problems subscore, gender, ID, speech impairment maternal education were then explored as possible factors affecting reporting of pain. Factors associated with pain severity were explored using a series of linear regression with pain severity scores (for the last day, week and 3 months) as the dependent variable, and using logistic regression with the absence/presence of moderate-severe regular pain as the dependent variable. Any factors indicating an association with pain severity at the p<.1 level were retained in a multivariate analysis which included GMFCS level as a co-variate to explore the significance of such factors over and above severity of CP. Logistic regression was also used to test whether the presence of pain at particular sites was associated with the above factors. A series of linear regressions examined the association between pain severity (entered as independent variable) and impact, as measured by the BPI (dependent variable), controlling for levels of GMFCS, ID and speech. *(see supplementary table A)* To examine the association between pain severity and quality of life, pairwise correlations were run between pain severity scores and K-10 domain t-scores, firstly within the PC data and then within the YP data. Concordance of reporting and level of agreement between PC and YP reports of pain and associated features, where both were available, were determined using intra-class correlation coefficients and Cohen's Kappa for continuous (pain severity scores) and categorical measures (absence/presence of regular pain, absence/presence of pain at various sites) respectively.

RESULTS PC pain questionnaires were completed for a total of 212 individuals, by mothers (n=185), fathers (n=17), foster carers (n=3), grandparents (n=2), sisters (n=2), carer (n=1), aunt (n=1) (respondent missing (n=1)). The YP were aged 13 years 8 months to 19 years 3 months (mean age = 16.7 years, SD 1.3) at the time. A total of 153 YP completed the self-report questionnaires. In 8 self-reporting YP, PCs said that they did not feel they could adequately describe the pain experiences of their teenager with typical learning ability so preferred not to comment. Both PC and YP questionnaires were completed for 145 YP. Rates of severe/profound ID were significantly higher in the group lacking YP reports and a higher proportion were non-verbal. Sample characteristics are presented in Table 1.

Table 1 here

Selective attrition in the whole cohort was previously estimated[25] comparing those who were seen at mean age 16 years with those who were not. Using GMFCS at age 2 years and

excluding those who had died, participants and non-participants did not differ in terms of GMFCS levels (p = .86).

Frequency and severity of PC reported pain

Seventy percent of PCs reported pain occurring in the last three months, 59% within the last week and 50% within the last day. Regular episodes of pain were reported by 56% of PCs, in the majority of whom this was moderate/ severe (89% of those with regular pain, 50% of the total sample).

Factors associated with PC reported pain

Pain was reported increasingly frequently with increasing levels of motor impairment, as indicated by GMFCS level and non-ambulant versus ambulant as well as in those with poorer general health at each time-period (all p values <.05 for pain for each of these predictors).

Table 2 presents frequencies of the various measures of pain occurrence by GMFCS level and for ambulant versus non-ambulant participants. PC report of pain (at any time-point) was not associated with level of deformity, intellectual disability, speech level or gender, SDQ score or maternal education (all p values > .07).

Table 2 here

Pain was associated with equipment use at each time period (all p values <.05), but this finding disappeared when level of motor impairment (GMFCS) was controlled for.

Factors associated with severity of PC reported pain

Results from the regression analyses are presented in Table 3. In the bivariate analyses, higher levels of GMFCS, equipment use, and general health problems were all associated with increased pain severity in the last day, week and 3 months (all p values<.05). The same was found for regular moderate/severe pain. The multivariate analyses produced significant regression equations for pain in the last day ($F(8, 200) = 3.96$, $p<.001$, $R^2=.14$), last week ($F(4, 204) = 5.02$, $p<.001$, $R^2=.09$), last 3 months ($F(5, 192)=3.83$, $p=.003$, $R^2=.09$), and for regular moderate/severe pain ($X^2(6)=23.7$, p<.001). The association between pain severity and general health remained (all p values <.001), even when GMFCS level was controlled for. When the general health index was replaced by a separate variable for constipation, the latter was associated with increased pain severity for the last day ($p=.02$) and 3 months ($p=.02$). Motor disorder type (specifically spasticity) was associated with increased pain severity in the last day ($p=.002$). PC report of moderate/severe regular

pain was associated with higher SDQ emotion scores (p=.02) and greater equipment use (p=.04).

Table 3 here

Location of pain. Table 4 presents frequencies of any pain reported by PCs at various sites in the last 3 months by GMFCS level. Pain in any of arm, back, hip, leg or chest was reported by 77% of PCs and 66% reported pain at multiple sites. PCs reported higher rates of hip [X^2 (1)=5.88, p<0.05] and abdominal pain [X^2 (1)=12.8, p<0.001] in the non-ambulant group compared to the ambulant group.

Table 4 here

A series of logistic regressions (*see Supplementary Table B*) tested whether pain at the various sites was associated with GMFCS level, deformity, general ill health (or specifically constipation) or gender. Hip pain was associated with higher levels of GMFCS (p=0.024) and constipation (p=0.015); abdominal pain was associated with higher levels of GMFCS (p=0.001), female sex (p=0.005), general ill health (p=.001) and constipation (p<0.001); chest pain was also associated with general ill health (p=.003) and constipation (p=0.008).

Convergence between PC and YP reports

For the 145 participants where both PC and YP reports were completed, parents reported pain a little more frequently than the YP, although not significantly so: 46% of PCs versus 42% of YP reported pain in the last day (one-sample proportion test, p = .42); 58% of PCs versus 50% of YP reported pain in the last week (p=.06); and 71% of PCs versus 63% of YP reported pain in the last 3 months (p = .05). However, intraclass correlations indicated good agreement between PC and YP pain severity scores (0 - 10) for pain occurring in the last day [ICC=0.60 (95% C.I. 0.48-0.69), p<0.001], the last week [ICC=0.70 (95% C.I. 0.61-0.78), p<0.001], and the last three months [ICC=0.75 (95% C.I. 0.66-0.82), p<0.001]. Regular pain was reported by 48% of YP of which the majority (82% of those with regular pain, or 39% of the total) was moderate to severe. Agreement between PC and YP self-reports of moderate-severe regular pain was good (k=0.69, 95% C.I. 0.56-0.79, p<0.0001). YP self-reports with PC reports were only available for 11 non-verbal YP, but within these there was strong agreement between PC and YP reports of moderate to severe regular pain: six individuals were reported by both PCs and the YP as experiencing moderate to severe regular pain, with just one YP reporting this level of pain when the PC did not (k =0.81, 95% C.I. 0.23-0.95, p=0.003).

Pain in any of arm, back, hip, leg or chest was reported by 71% of YP. Hip pain was associated with female sex (p=0.024); and abdominal pain associated with constipation

(p=0.020). Agreement between PCs and YP reports of pain at each site was fair to good (kappa values ranged 0.48-0.64, all p<0.0001). Almost two thirds (63.4%) of PCs reported pain at multiple sites, as did a similar proportion (61.4%) of YP, with moderate agreement as to which individuals this applied to (k =0.52, 95% C.I. 0.34-0.64, p<0.0001).

Timing of pain

Parents and YP were asked when pain occurred and was worst. The most frequent response was during voluntary movement, both in PC reports (41%) and YP reports (67%) [X^2 (1)=9.72, p<0.05)]. This was followed by when moved (25% in both PC and YP reports), and then during daytime rest (17% in both PC and YP reports). Pain during voluntary movement was associated with lower GMFCS levels (as a continuous variable) [β= -0.44, SE=0.14, p=0.003]; pain when moved was associated with higher GMFCS levels [β=1.05, SE=0.24, p<0.0001]. PCs tended to report pain was worst at night more frequently than YP 28% versus 7%, [X^2(1)=3.44, p=0.06]. Pain "all the time" was reported by 11% of PCs and 3% of YP (Fisher's exact, p=0.12). Pain was reported when eating by 2 individuals.

Medication and other interventions for pain

112 (53%) PCs and 74 (48%) YP reported their use of analgesic, anti-spasticity and other medicines.

Simple analgesics (e.g., paracetamol and ibuprofen) were used by 77% of responding YP – frequency = 'often' or 'very often'. PCs reported use of simple analgesics in 88% of the YP – 'often' or 'very often'.

Muscle relaxants were the next most frequently used agents that may impact on the pathophysiology of pain: Baclofen (14%), Diazepam (6%) and regular Botulinum Toxin A injections (11%) as well as anti-reflux medications (12%) and laxatives (14%).

PCs reported a third of YP were on more than 2 medications that may impact on Pain. 3% of families regularly used alternative therapies such as Acupuncture, Osteopathy and Homeopathy.

Quality of life for YP and the impact of pain

The modified Brief Pain Inventory Scale was completed by 147 YP explicitly describing the impact of pain on general activity, recreational and social activity, relationships, school/college/work, mobility, self-care, sleep, mood and overall enjoyment of life. Mobility was the area most affected (28% "quite a lot/a great deal"), followed by mood (20%), general activity (17%) and recreational activity (16%). Total impact scores were found to be significantly predicted by PC pain severity scores for the last day (β=1.67, SE=0.20,

p<0.001) past week, (β=1.76, SE=0.17, p<0.001)and 3 months (β=1.37, SE=0.16, p<0.001); also for YP pain severity scores for the last day (β=.1.64, SE=.20, p<.001) past week (β=1.55, SE=0.15, p<0.001), and 3 months (β=1.46, SE=0.14, p<0.001). Levels of GMFCS, ID and speech were not associated with total impact scores (all p-values>0.2, see Supplementary Table A).

The Kidscreen was completed by 187 PCs and 129 YP. Pain severity reported by PCs in the last week *and* in the last 3 months were negatively correlated with physical well-being domain t-scores [r (184)= -0.31, p<0.0001; r (183)= -0.36, p<0.001] and psychological well-being t-scores [r (183)= -0.01, p=0.01]; [r(183)= -0.17, p=0.02]; and in the last 3 months, with mood and emotions t-scores [r (181)= -0.16, p=0.03], but no other domains. YP reports of pain severity in the last week and 3 months were negatively correlated with YP physical wellbeing t-scores [r(126)= -0.31, p<0.001]; [r(126 = -.40, p<0.001], mood and emotions t-scores [r(126)= -0.21, p=0.02)] and with psychological well-being t-scores [r(126)= -0.31, p<0.001] and autonomy t-scores [r(126)= -0.20, p=0.02] for pain in the last 3 months.

Discussion

Our study of surviving teenagers with bilateral cerebral palsy from a total population cohort confirms that pain is a common experience for these young people at an age when YP are striving for greater independence in the world[28,] and is associated with severity both of motor impairment (increasing GMFCS and non-ambulant) and general health but not deformity, gender, SDQ score, intellectual disability or maternal education (this is in line with previous cross sectional studies)[3,4,6] . It is important to note that some YP in all GMFCS groups experience regular moderate/severe pain.

A key finding from this study is that severity of general ill-health, independent of motor severity, is the factor most strongly associated with pain severity at all time periods and with regular moderate/severe pain. Gastrointestinal dysfunction in CP especially problems of motility, for which constipation is a marker, has received more attention in recent years but management remains a challenge. For many YP GI function remained a problem despite treatment. Spasticity was associated with greater pain severity in the last day and moderate/severe regular pain with the use of postural modifying equipment and a higher SDQ emotional score, as found previously [4,14].

Parent/carer and YP agreement both for pain presence and severity was high (Kappa of .69 for moderate/severe regular pain, agreement previously reported as Spearman rank 0.45 [3,4]) and fair to good for site mainly arm, back, hip, leg or chest. Abdominal pain was more common in females. Two thirds reported multiple sites of pain.

Our prevalence of pain rates, 70% in the last three months, are similar to those reported in the SPARCLE study of similar age[4], although that study included teenagers with unilateral as well as bilateral cerebral palsy. Reported rates of pain in a typical adolescent population are also high, 74% experiencing pain in any of headache, backache or stomach ache at least monthly in the last 6 months, more in females than males which differs from our study[29]. However, in our study 56% of YP experienced regular pain, moderate or severe in 89%, despite significant medication use.

Pain severity had a negative impact on quality of life assessed using the BPI which explicitly links pain to impact and although the Kidscreen does not make a direct link to pain, the BPI and Kidscreen findings were very similar in measures of physical and psychological wellbeing.

Pain in CP has previously been linked to physical interventions including those intended to be therapeutic[4]. We found that pain occurs at different times and with different activities in different GMFCS groups. Pain was reported in our non-ambulant group when being moved and in the ambulant group when actively mobile and in the chest/shoulders and arms of children using assistive mobility device. An important factor to be considered in clinical care is our finding that therapeutic postural modifying equipment independent of severity of motor impairment was associated with moderate/severe regular pain. An inability to move independently often means pain becomes more obvious at night but this was reported more by parents than YP and there was no correlation with perceived impact on quality of life and sleep from YP self-report. Of course, a potential confounder is that the most severely impaired group were less able to comment on their sleep quality.

While self-report remains the gold standard when assessing pain, extrapolation from these findings allows reasonable confidence to be assumed for PC report as a proxy measure.

Recognition is only half the battle. As stated by Blackman et al[18], recognition of Pain is step one in our understanding of its underlying pathophysiology in YP with CP. In turn this may aid us in focussing on appropriate and potentially novel ways of pharmacological and therapeutic intervention to enable comfort and function.

Summary, review and recommendations

Increasing awareness of pain and the co-morbidities in CP, particularly general health and constipation, may help prevent and more effectively treat and thereby reduce the high levels of pain experienced by young people with cerebral palsy. In line with the recommendations

of the recent NICE guidelines it is imperative to consider the potential mechanisms and the physical and emotional manifestations of Pain in any YP with CP.

- Pain is a common, significant experience in teenagers with bilateral Cerebral palsy
- Appropriate recognition and management of pain is at the core of any individual's clinical need.
- Unless core Physical and Psychological comfort is optimised any positive impact of more complex multidisciplinary interventions to improve function in YP with CP can be compromised.
- If the functional ability of any individual with CP regresses, then it is important to consider Pain as a potential root cause.
- Before any new clinical medical or therapeutic intervention is considered, it is important to consider the potential impact that it may have on Pain in the Young person with CP.
- As such it is vital that appropriate, evidence based Pain scores should be used as an integral part of regular reviews of YP with CP, in order to guide recognition and management by all individuals involved in their care.

Strengths of study are a total geographically derived population cohort with *bilateral* Cerebral Palsy; both parent and teenager reports with measures of frequency, severity, site, time, impact and intervention for pain and a range of associated factors.

Limitations of the study are that this is across sectional questionnaire study, there are small numbers in some YP self-reporting GMFCS sub-groups and incomplete reporting in questionnaires by a small number of YP.

Acknowledgments: We would like to thank all the families, and especially the young people, for their participation; Anne Greany, Alison Davis and Elspeth Will for data and Lewis Rosenbloom for wise counsel.

Contributor's Statement: Gillian Baird, Charlie Fairhurst and David Scrutton conceived and designed the study. Emily Simonoff advised on analysis; Susie Chandler, Adam Shortland, Gillian Baird and Charlie Fairhurst analysed and interpreted the data. Gillian Baird and Charlie Fairhurst drafted the article and all authors revised it critically. Susie Chandler had full access to all of the data (including statistical reports and tables) in the study and with Emily Simonoff takes responsibility for the integrity and accuracy of the data analysis. All

authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Potential Conflicts of Interest: The authors have no conflicts of interest relevant to this article to disclose.

Funding source: The study was funded by the Charles Wolfson Charitable Trust, Cerebra, One Small Step and the Hickman Fund (Guy's & St Thomas' Charity). Emily Simonoff receives support from the National Institute for Health Research (NIHR) Mental Health Biomedical Research Centre at South London and Maudsley NHS Foundation Trust and King's College London.

The South-East MREC granted ethical approval, reference number 06/MRE01/18

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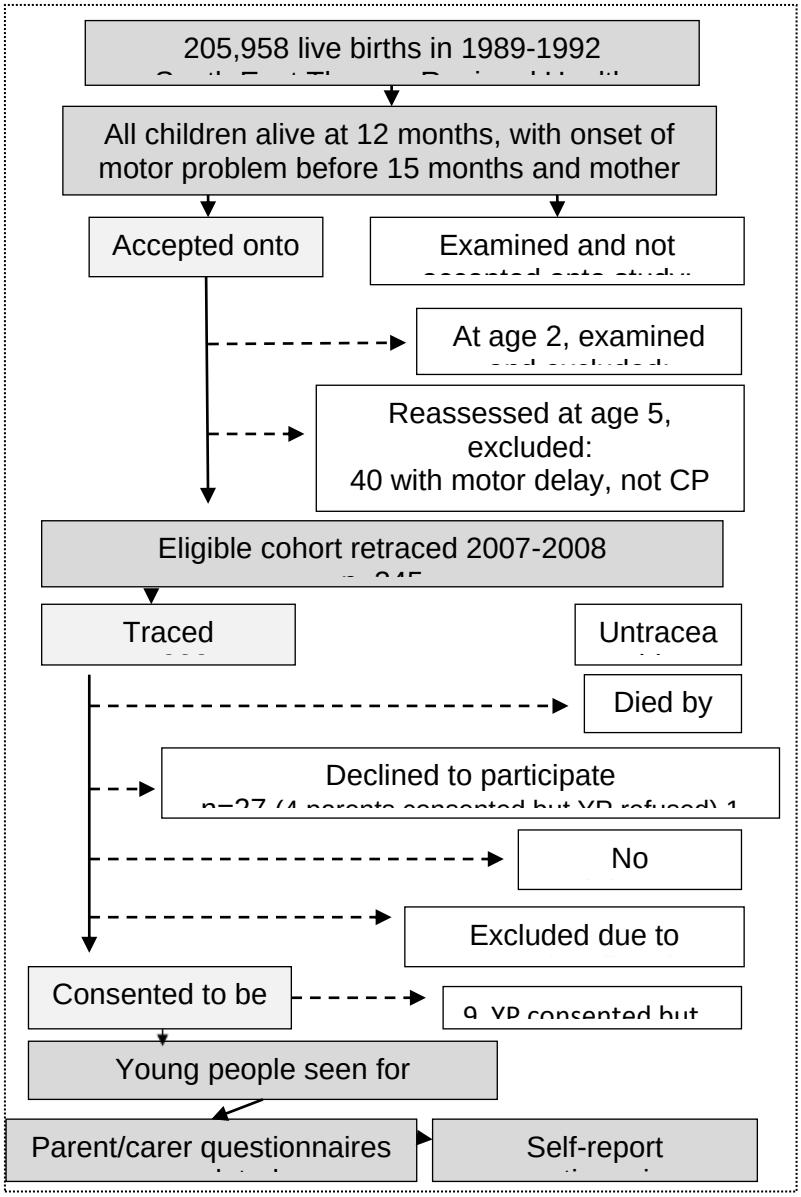

Figure 1: Sample Ascertainment

Table 1. Participant characteristics

Characteristics	PC data (N=212)	PC and YP data (n=145)	PC data only (n=67)
Male (n (%))	132 (62.3)	97 (66.9)	35 (52.2)
Female (n (%))	80 (37.7)	48 (33.1)	32 (47.8)
Mean age (SD)	16.7 years (1.3)	16.8 (1.2)	16.6 (1.3)
Gestational age (n (%)):			
>40 weeks (post term)	3 (1.4%)	3 (2.1%)	0 (0%)
37-40 weeks (term)	107 (50.5%)	63 (43.5%)**	44 (65.7%)**
32 – 36 weeks (mod preterm)	39 (18.4%)	27 (18.6%)	12 (17.9%)
28 – 31 weeks (very preterm)	38 (17.9%)	32 (22.1%)*	6 (9.0%)*
<28 weeks (extremely preterm)	25 (11.8%)	20 (13.8%)	5 (7.5%)
GMFCS I (n (%))	27 (12.7)	26 (17.9)***	1 (1.5)***
GMFCS II (n (%)	58 (27.4)	50 (43.5)***	8 (11.9)***
GMFCS III (n (%))	27 (12.7)	25 (17.2)**	2 (3.0)**
GMFCS IV (n (%))	34 (16.0)	22 (15.2)	12 (17.9)
GMFCS V (n (%))	66 (31.1)	22 (15.2)***	44 (67.7)***
Ambulant (n (%))	133 (62.7)	111 (76.5)	22 (32.8)
Non-ambulant (n (%))	79 (37.3)	34 (23.5)***	45 (67.2)***
Primary disorder, n=210:			
spastic (n (%)	164 (78.1)	113 (78.5)	51 (77.2)
dystonic (n (%))	18 (8.6)	11 (7.6)	7 (10.6)
ataxia (n (%))	11 (5.2)	9 (6.3)	2 (3.0)
involuntary athetosis (n (%))	4 (1.9)	2 (1.4)	2 (3.0)
other (n (%))	13 (6.2)	9 (6.3)	4 (6.1)
Constipation problems (n (%))	75/210 (35.7)	45 (31.3)*	30 (45.5)*
GI reflux/upper GI problems (n (%))	26/211 (12.3)	10 (6.9)***	16 (23.9)***
Mean health index (SD), n=211	1.78 (1.78)	1.37 (1.57)***	2.64 (1.91)***
Mean lower limb deformity score (SD), n=183	6.0 (4.15)	7.39 (4.11)***	5.17 (3.33)***
Speech			
Normal/typical (n (%))	77 (36.3)	73 (50.3)***	4 (6.0)***
Mainly intelligible to unfamiliar (n (%))	49 (23.1)	42 (29.0)**	7 (10.5)**
Mostly not intelligible to unfamiliar (n (%))	21 (9.9)	14 (9.7)	7 (10.5)
Severe but mostly familiar to familiar (n (%))	8 (3.8)	5 (3.5)	3 (4.5)
Open vowels only, unintelligible to all (n (%))	57 (26.8)	11 (7.6)***	46 (68.7)***
Level of intellectual disability, n=211			
None (n (%))	45 (21.3)	45 (31.0)***	0 (-)***
Mild (n (%))	44 (20.9)	42 (29.0)***	2 (3.0)***
Moderate (n (%))	44 (20.9)	36 (24.8)*	8. (12.1)*
Severe (n (%))	50 (23.7)	19 (13.1)***	31 (47.0)***

Profound (n (%))	28 (13.3)	3 (2.1)	25 (37.8)***
Ethnicity: White Caucasian (n (%))	174 (82.5)	119 (82.6)	55 (82.1)
Maternal Education, n=201:			
No formal qualifications (n (%))	38 (18.9)	26 (18.4)	12 (20.0)
GCSEs (n (%))	78 (38.8)	52 (36.9)	26 (43.3)
A-levels (n (%))	24 (11.9)	19 (13.4)	5 (8.3)
City and Guilds (n (%))	26 (12.9)	19 (13.5)	7 (11.7)
Undergraduate degree (n (%))	29 (14.4)	20 (14.2)	9 (15.0)
Postgraduate degree (n (%))	6 (3.0)	5 (3.6)	1 (1.7)
Employment: at least one parent employed (n (%))	106/204 (52.0)	66 (46.2)*	46 (65.6)*
N (%) reporting pain in time period:			
Last day	105 (50.4)	67 (46.2)	38 (56.7)
Last week	126 (59.4)	84 (57.9)	42 (62.7)
Last 3 months	149 (70.3)	103 (71.0)	46 (68.7)
N (%) with moderate-severe regular pain	106 (50.0)	70 (48.3)	36 (53.7)
Equipment use:			
Prone-standing frame (n (%))	17 (8.0)	4 (2.3%)**	13 (19.4)**
Upright standing-frame (n (%))	36 (17.0)	16 (11.0)**	20 (29.9)**
Night lying support (n (%))	35 (16.5)	16 (11.0)*	19 (28.4)*
Spinal brace (n (%))	11 (5.2)	1 (0.7)***	10 (14.5%)***
Hip abduction brace (n (%))	2 (0.9)	-	2 (3.0)
Ankle-foot-orthotics (n (%))	45 (21.2)	28 (19.3)	17 (25.4)
Knee gaiters (n (%))	18 (8.5)	10 (6.9)	8 (11.9)

*Fisher's Exact or X^2, p<.05, **Fisher's Exact, X^2, p<.01, ***Fisher's Exact, X^2, or t-test p<.001

Table 2. Parent-reported pain frequency (%) by GMFCS level for pain experienced in last day, last week and last three months and regular mod/severe pain

	GMFCS Level					Test of trend p-value		
	I	II	III	IV	V		Ambulant	Non-ambulant
Pain in last day	5 (19.5%)	27 (46.6%)	15 (55.6%)	18 (52.9%)	40 (60.6%)	.001	57 (42.9%)	48 (60.8%)
Pain in last week	11 (40.7%)	33 (56.9%)	17 (63.0%)	21 (61.8%)	44 (66.7%)	.036	70 (52.6%)	56 (70.9%)
Pain in last 3 months	14 (51.9%)	38 (65.5%)	20 (74.1%)	25 (73.5%)	52 (78.8%)	.010	83 (62.4%)	66 (83.5%)
Moderate/severe regular pain	8 (29.6%)	26 (44.8%)	16 (59.3%)	19 (55.9%)	37 (56.1%)	.024	58 (43.6%)	48 (60.8%)

Table 3: Regression coefficients for potential predictors of PC reports of pain severity, and moderate/ severe regular pain

Timescale	Covariate	Bivariate regression Coefficient (SE)	p-value	Multivariate regression Coefficient (SE)	p-value
Pain in last day (0-10)	Gender	-.24 (.34)	.49		
	GMFCS level	.28 (.11)	.01	.09 (.17)	.60
	Motor type	*-1.8 (.59)*	*.003*	*-1.97 (.59)*	*.002*
	Deformity score	.05 (.04)	.19		
	Equipment use	.45 (.18)	.01	.16 (.23)	.49
	General health index	*.37 (.09)*	*<.001*	*.32 (.10)*	*.002*
	Constipation	*.1.25 (.34)*	*<.001*	*.87 (.35)*	*.02*
	SDQ total	.08 (.05*)*	.89		
	SDQ emotion	.12 (.10)	.22		
	Intellectual disability	.07 (.09)	.42		
	Speech scale	.18 (.10)	.09	-.03 (.15)	.81
	Maternal education	.18 (.12)	.13		
Pain in last week (0-10)	Gender	-.04 (.37)	.92		
	GMFCS level	.27 (.12)	.03	-.07 (.16)	.67
	Motor type	-1.91 (.64)	.003	-.17 (.11)	.12
	Deformity score	.02 (.05)	.66		
	Equipment use	.62 (.19)	.002	.40 (.25)	.12
	General health index	*.38 (.10)*	*<.001*	*.30 (.11)*	*.005*
	Constipation	.99 (.38)	.009	.63 (.39)	.12
	SDQ total	.01 (.05)	.83		
	SDQ emotion	.19 (.11)	.38		
	Intellectual disability	.04 (.10)	.69		
	Speech scale	.10 (.11)	.38		
	Maternal education	.15 (.13)	.25		
Pain in last 3m (0-10)	Gender	-.05 (.42)	.91		
	GMFCS level	.36 (.14)	.01	.14 (.18)	.44
	Motor type	-1.47 (.75)	.05	-.01 (.13)	.92
	Deformity score	.04 (.05)	.41		
	Equipment use	.48 (.22)	.03	.12 (.29)	.68
	General health index	*.45 (.11)*	*<.001*	*.34 (.12)*	*.006*
	Constipation	*1.43 (.42)*	*.001*	*1.07 (.44)*	*.02*
	SDQ total	.06 (.06*)*	.33		
	SDQ emotion	.18 (.12)	.13		
	Intellectual disability	-.02 (.11)	.81		
	Speech scale	.17 (.12)	.16		
	Maternal education	.27 (.14)	.07	.25 (.14)	.08
Moderate/severe regular pain (absent/present)	Gender	.16 (.28)	.57		
	GMFCS level	.22 (.10)	.03	-.24 (.25)	.33
	Motor type	-.97 (.52)	.07	-1.3 (1.04)	.30
	Deformity score	.06 (.04)	.17		
	Equipment use	.55(.16)	<.001	.84 (.41)	.04
	General health index	.31 (.09)	<.001	-.07 (.15)	.63
	Constipation	.51 (.29)	.08	-.56 (.61)	.35
	SDQ Total	.07 (.05)	.17		
	SDQ emotion	*.21 (.10)*	*.03*	*.32 (.13)*	*.02*
	Intellectual disability	.10 (.08)	.21		
	Speech scale	.13 (.09)	.12		
	Maternal education	.23 (.10)	.03	-1.31 (1.27)	.30

Table 4. Frequencies (%) of PC reports of pain by site and GMFCS, and PC-YP agreement on reports of pain at each site

	GMFCS level					Test of trend p-value	PC-YP agreement on site of pain (k (95% CI))
	I	II	III	IV	V		
	n=27	n=58	n=27	n=34	n=66		
Leg	15 (55.6%)	33 (56.9%)	19 (70.4%)	18 (52.9%)	31 (48.0%)	.272	.60 (.45-.71), p<.0001
Hip	7 (25.9%)	15 (25.9%)	17 (63.0%)	18 (52.9%)	33 (50.0%)	.002	.55 (.40-.68), p<.0001
Back	8 (9.5%)	22 (26.2%)	12 (44.4%)	10 (29.4%)	32 (49.2%)	.140	.59 (.44-.70), p<.0001
Abdomen	6 (22.2%)	16 (27.6%)	11 (40.7%)	18 (52.9%)	40 (60.6%)	<.001	.51 (.35-.64), p<.0001
Chest	4 (14.3%)	4 (6.9%)	5 (18.5%)	2 (5.9%)	13 (20.0%)	.206	.48 (.27-.65), p<.0001
Head	10 (27.0%)	17 (29.3%)	8 (29.6%)	6 (17.7%)	24 (36.9%)	.920	.55 (.39-.68), p<.0001
Arm	6 (22.2%)	14 (24.1%)	8 (29.6%)	5 (14.7%)	19 (29.2%)	.647	.64 (.47-.76), p<.0001

Supplementary Table A. Regression coefficients for predictors of total impact score from the Brief Pain Inventory

	Covariate	Coefficient (SE)	p-value
Total impact	*Pain in the last day (PC report)*	*1.67 (.17)*	*<.001*
	GMFCS level	.06 (.42)	.880
	Speech scale	.02 (.49)	.98
	Intellectual disability	-.56 (.44)	.20
	Pain in the last week (PC report)	*1.76 (.17)*	*<.001*
	GMFCS level	.09 (38)	.80
	Speech scale	.21 (.45)	.65
	Intellectual disability	-.54 (.40)	.18
	Pain in the last 3 months (PC report)	*1.37 (.16)*	*<.001*
	GMFCS level	-.10 (.42)	.81
	Speech scale	-.24 (.48)	.63
	Intellectual disability	-.54 (.44)	.22
Total impact	*Pain in the last day (YP report)*	*1.64 (.20)*	*<.001*
	GMFCS level	.12 (.41)	.77
	Speech scale	-.12 (.48)	.80
	Intellectual disability	-.30	.49
	Pain in the last week (YP report)	*1.54 (.15)*	*<.001*
	GMFCS level	.04 (.38)	.92
	Speech scale	.10 (.33)	.83
	Intellectual disability	-.42 (.40)	.29
	Pain in the last 3 months (YP report)	*1.46 (.14)*	*<.001*
	GMFCS level	.16 (.37)	.68
	Speech scale	-.60 (.43)	.17
	Intellectual disability	-.26 (.39)	.50

Supplementary Table B. Regressions coefficients for potential predictors of pain at various sites

	Covariate	Coefficient (SE)	p-value
Leg pain	GMFCS level	-.21 (.15)	.14
	Deformity score	.02 (.05)	.74
	General health index	.09 (.09)	.34
	Constipation	.17 (.33)	.62
	Gender	.37 (.32)	.26
Hip pain	*GMFCS level*	*.54 (.15)*	*.02*
	Deformity score	.05 (.05)	.31
	General health index	.14 (.09)	.14
	Constipation	*.91 (.35)*	*.02*
	Gender	.63 (.32)	.05
Back pain	GMFCS level	.06 (.15)	.71
	Deformity score	-.001 (.05)	.98
	General health index	.09 (.09)	.28
	Constipation	.67 (.34)	.06
	Gender	.24 (.32)	.46
Abdominal pain	*GMFCS level*	*.21 (.16)*	*.001*
	Deformity score	.04 (.06)	.45
	General health index	*.38 (.10)*	*.001*
	Constipation	*1.53 (.37)*	*<.001*
	Gender	*1.05 (.35)*	*.005*
Chest pain	GMFCS level	-.02 (.23)	.90
	Deformity score	-.02 (.09)	.85
	General health index	*.38 (.13)*	*.003*
	Constipation	*1.14 (.49)*	*.01*
	Gender	.34 (.49)	.49
Pain in the Head	GMFCS level	.65 (.16)	.68
	Deformity score	-.12 (.07)	.09
	General health index	.24 (.10)	.13
	Constipation	.74 (.35)	.36
	Gender	.17 (.34)	.62
Arm pain	GMFCS level	-.19 (.17)	.29
	Deformity score	.06 (.06)	.33
	General health index	.13 (.10)	.10
	Constipation	.55 (.38)	.15
	Gender	.60 (.37)	.12

Pain Questionnaire SH&PE

(parent report)

Please put a point on the line that fits best with your child's pain.

1. Over the past day… Have they experienced any pain?

0 No pain 1 2 3 4 5 6 7 8 9 10 Pain as bad as it could be

2. Over the last week… Have they experienced any pain?

0 No pain 1 2 3 4 5 6 7 8 9 10 Pain as bad as it could be

3. Over the last three months… Have they experienced any pain?

0 No pain 1 2 3 4 5 6 7 8 9 10 Pain as bad as it could be

4. In which place (s) does your child get pain? *(Please circle the best answer)*

Place		Not at all	A little	Quite often	All the time
Legs	Left	0	1	2	3
	Right	0	1	2	3
Hips	Left	0	1	2	3
	Right	0	1	2	3
Back		0	1	2	3
Tummy		0	1	2	3
Chest		0	1	2	3

Head		0	1	2	3
Arms	Left	0	1	2	3
	Right	0	1	2	3
Other (Please specify)		0	1	2	3
Other (Please specify)		0	1	2	3

5. Does your child experience pain *regularly*? ☐ **Yes** ☐ **No**

6. Please *circle the word* that best describes the severity of this *regular* pain.

1**Mild** 2**Moderate** 3**Severe**

7. If they do experience pain *regularly*, when is that? *(Please mark one or more if applicable)*

1☐ In the daytime at rest

2☐ In the day-time with voluntary movement

3☐ When someone moves them

4☐ At night-time

5☐ All the time

6☐ When eating or talking

7☐ Other

Which time is worst? ……

Appendix

Deformity Index

The passive range of each joint was assessed using a goniometer. This was performed in the supine position for the lower limb. To detect flexion contractures, the pelvis was stabilised and the contralateral hip was flexed to end range and the angle between the surface the participant was lying on and the thigh was measured. Hip rotation measures were taken with the hip and knee flexed to 90º where possible. In instances where there was a restriction to either hip or knee flexion, the lower limb joints were flexed to just short of full available flexion and rotation assessed in this position. Varus and valgus foot deformity and hallux valgus were noted and calculated as present or absent.

An index of deformity was developed based on that used by Ostensjo 2004. Each joint range of movement was scored as 0 or 1 based on whether the range fell more than 2 SD outside the normal. The lower limb ranges were based on normal range identified from a group of 14 normally developing adolescents assessed in this hospital (age 13-22 mean 16.6yrs, 7 females 7 males). The sum of index components on each side of the body can reach a maximum of 14. Scores for both sides of the body are summed and added to this scale is a spinal score. It is difficult to calculate a measure for the spine from clinical assessment, so the following scoring from clinical findings was developed: a YP may have both sagittal plane and coronal plane changes. It was hypothesised that surgical fixation of the spine was a different but still significant change in body ROM and so was scored as noted.

REF Ostensjo 2004

Spine:

0 = Neutral

1 = Sagittal plane changes

1 = Coronal plane changes

1 = Surgical correction

1 = Fixed pelvic obliquity in sitting

Lower limb

Score	1	2	3		1SD	2SD	3SD
Hip flexion	**<120**	**<100**	**<90**	Score	1	2	3
Hip extension	**<0**	**<-20**	**>-20**	Hip flexion	**<120**	**<100**	**<90**
Hip abduction (flexion)	**<45** >70	**<30** >80	**<20** >95	Hip extension	**<0**	**<-20**	**>-20**
Hip internal	**<35**	**<25**	**<15**	Hip abduction	<43	<30	<17

rotation	>55	>65	>75	(flexion)	>69	>81	>94
Hip external rotation	**<35** >60	**<25** >75	**<10** >85	Hip internal rotation	<35 >54	<26 >64	<16 >74
Knee extension	**>1 FFD** > 5 hyper	**>5 FFD** > 10 hyper	**>10 FFD** >15 hyper	Hip external rotation	<35 >61	<24 >73	<11 >86
Popliteal angle	**>60**	**> 70**	**>80**	Knee extension	>1 FFD > 5 hyper	>5 FFD > 10 hyper	>10 FFD >15 hyper
Ankle dorsiflexion	**<10DF** > 20DF	**<0º DF** >25 DF	**>5PF** >30 DF	Popliteal angle	>60	> 70	>77
				Ankle dorsiflexion	> 20DF <9DF	>25.5 DF <3º DF	>31DF >2PF

1

2 Valgus/varus foot deformity no=0, mild =1, significant=2